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Psychosocial functioning in siblings of paediatric cancer patients one to six months after diagnosis

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Abstract

The aim of this study was to prospectively investigate the prevalence of and risk factors for psychosocial problems in siblings of paediatric cancer patients. One and 6 months after diagnosis, sibling self-reported anxiety, social–emotional problems and quality of life (QoL) were assessed, as were the predictor variables: sibling prediagnosis functioning, age and gender and the ill child's diagnosis. At 1 month, siblings reported a lower QoL and adolescent girls reported more emotional problems compared with peers. At 6 months, adolescent QoL remained relatively impaired. Over time, adolescent brothers reported fewer emotional and total problems and young girls reported decreased anxiety. No significant amelioration in QoL was found over time. The older the siblings were, the lower their observed QoL at both measurements and in several domains. The occurrence of life events predicted sisters' QoL at 1 month. Changes in sibling functioning were predicted by none of the investigated risk factors. Thus, QoL is impaired shortly after diagnosis. Adolescent siblings risk persisting problems in daily functioning. Further prospective research on other risk factors such as coping and family functioning over time is needed.

Keywords: Paediatric cancer; Siblings; Social–emotional functioning; Quality of life; Risk factors; Prospective study design

1. Introduction

When a brother or sister is diagnosed with cancer, siblings experience intrusive changes in family life. Siblings of paediatric cancer patients need to adapt to these changes and to the intrusive emotions such as fear, anger, isolation, jealousy, shame and guilt, which may be related to the illness of their ill sibling. They have to adjust to changes in family routines, to increased responsibilities and often to a decreased physical and emotional availability of the parents. The literature presents contradictory evidence for the risk of the development of psychosocial problems among siblings after the diagnosis of cancer in their brother or sister. Significant internalising problems, such as emotional and social withdrawal, anxiety, feelings of guilt, hopelessness, shame and sadness; or externalising problems, such as anger, non-compliance or other acting-out behaviour, were reported in several studies [1–20]. However, others found no substantial social–emotional problems in siblings of paediatric cancer patients [21–26]. Contradictory results are due to differences in samples, in study designs, and in the assessment and conceptualisation of psychological adjustment [27]. The paediatric oncologist and the nurse are the first professionals to encounter adjustment problems in family members of a child with cancer. It is most important that adjustment problems in siblings are recognised by medical staff at an early stage. Early recognition of adjustment problems is possible only when we know which children are 'at risk' for psychosocial problems.

Firstly, it can be argued that children who are vulnerable before the diagnosis of cancer in a sibling are at risk of developing more serious problems after the diagnosis [18,28]. Two studies indicate such a relationship between previous problems and later adjustment.

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Fife and colleagues [7] conducted a longitudinal study comprising the whole family of a child with cancer. They studied the psychosocial impact of the illness on 33 families. They found that in those families where problems existed prior to the diagnosis, family life deteriorated and family members had difficulties coping with the illness. In a quantitative study by Sahler and colleagues on sibling adjustment, pre-existing problems and postdiagnosis functioning were assessed systematically in 254 siblings. The results indicated that pre-existing problems were a major risk factor for subsequent problems in siblings of children in different phases of their treatment for cancer [16]. The sibling's previous functioning may thus be recognised as an important factor in the early recognition of later adaptational problems, but the aforementioned results have not yet been replicated in other studies.

Demographic factors and illness characteristics may also indicate why some siblings are more at risk for psychosocial adjustment problems than others. Again, study results are contradictory. Several studies found no differences between boys and girls in psychosocial adjustment to the illness [1,6,18,24]. Other studies found differences according to sibling age and gender. Young boys [16,29] and adolescent siblings [29] (adolescent sisters in particular [16]) were at risk for increased emotional distress. Furthermore, several studies have demonstrated that younger siblings either experience more adverse effects [10,17,30] or a more limited positive growth [1,5] than older siblings. In another study, no age differences were found regarding good or poor adjustment to the illness [18]. Differences in the nature of problems experienced at different ages were reported as well. Lower self-esteem was found in siblings aged 4–6 years, while depressive symptoms and anxiety were more pronounced in siblings aged 7–12 years [19]. In another study, siblings aged 8–13 years experienced feelings of isolation and loneliness, fear of becoming sick themselves and anger, whereas the older adolescent siblings experienced more complex feelings such as guilt, burden from their sense of responsibility for the ill child, and ambivalence towards their ill sibling [2]. These studies indicate that both younger and adolescent siblings seem to be at risk for adjustment problems, but these may be of a different nature. The differences according to age and gender are complex; they require a developmental viewpoint and need to be studied more thoroughly.

Besides the characteristics of the sibling, characteristics of the illness should also be considered. The diagnosis, treatment, number of hospital visits and side-effects will determine the burden of the illness for all family members. In a small sample, Madan-Swain and colleagues [24] found an association between the sibling's coping styles and the ill child's diagnosis. Siblings of a child that was diagnosed with a solid tumour seemed to engage more in self-oriented introspective strategies than siblings of children that were diagnosed with other forms of cancer. It was argued that, as a consequence of a relatively short treatment period, these siblings would have less access to information or support, and would have to rely more on their own cognitive coping abilities than be able to express their emotions or seek support. The number of nights that were spent in hospital was found to predict adjustment problems in siblings in a study by Sloper and While [18]. The disruptiveness of the illness for family life, in terms of hospital visits, thus seems to affect the sibling's wellbeing. The illness may also burden the sibling with extra responsibilities. Worries about the ill sibling may stimulate siblings to care for or be more considerate towards the ill child. For example, Barbarin [31] found that siblings became more independent the more severe the illness was. Two studies failed to demonstrate any effect of illness variables on the existence of problematic behaviour in the sibling [1,16].

Besides the prevalence of adjustment problems in different subgroups of siblings, adjustment should be regarded as a process. The question is how sibling adjustment changes/develops over time. In a cross-sectional study, parents reported fewer internalising and externalising problems when more time had elapsed since the diagnosis [6]. In a single prospective study that was performed on sibling adjustment, it appeared that the prevalence of psychosocial problems did not diminish over time. Problems of a different nature were reported that differed according to the phase of the illness [17]. Psychosocial problems in siblings improved slightly during remission of the illness. However, anxiety remained relatively high during all phases of the illness in most of the siblings.

In summary, the aforementioned study results suggest a relationship between sibling demographic characteristics, sibling prediagnosis functioning, and illness characteristics on the one hand, and the prevalence and persistence of psychosocial problems after the ill child's diagnosis of cancer on the other hand. Nevertheless, consistent evidence on the nature of the psychosocial problems and on risk factors is lacking. Considering the lack of longitudinal studies, it becomes evident that the course of sibling psychological adjustment over time should be investigated further.

The first aim of the present prospective study was to investigate the nature and prevalence of psychosocial problems in siblings of paediatric cancer patients during the first 6 months following diagnosis. The second aim was to determine the contribution of different risk factors that exist prior to diagnosis, such as sibling age, gender, physical complaints, healthcare use and life-events and of illness characteristics, to sibling functioning over time.

2. Patients and methods

2.1. Participants

Families of children diagnosed with cancer were recruited from two divisions of paediatric oncology, firstly at the Emma Children's Hospital in the Academic Medical Centre in Amsterdam, and secondly at the University Hospital in Groningen. Inclusion criteria were: a first diagnosis of cancer in the ill child; a maximum of 4–8 weeks between the medical diagnosis and recruitment for the present study; the presence of siblings aged 7–18 years in the family; and sufficient command of the Dutch language by parents and siblings. A maximum of two siblings per family was included in order to prevent an overrepresentation of larger families. Seventy-one families were eligible, considering the inclusion criteria of language and sibling age. Of these families, 56 agreed to participate (79%). This first measurement (M1) group consisted of 83 siblings aged 7–18 years, 37 boys (45%) and 46 girls (55%).

The ill children had different diagnoses of cancer (Table 1). The patients were all treated according to European standards. Of the children with leukaemia, one child had a bone marrow transplantation. Of the children with lymphoma, eight were treated with chemotherapy alone, three were treated with chemotherapy and surgery, and one child with chemotherapy and radiotherapy. Of the children with a solid tumour, 11 were treated with chemotherapy and surgery, eight had chemotherapy, radiation and surgery, five had chemotherapy alone and two children had surgery alone. Three of the children with a solid tumour underwent an amputation. Of the children with a brain tumour, one child had radiotherapy and surgery, three had chemotherapy alone and one child had chemotherapy and surgery. The age of the ill children ranged from 1 to 16 years (mean age 9 years; S.D. 4.4), 35 (63%) were boys and 21 (38%) were girls.

Fathers and mothers were asked to participate in the interview alternately but, in most of the families (67%), the mothers were interviewed for practical reasons.

The 15 families that refused participation did not differ significantly from the participants with regard to sibling age ($t = 0.63$; degrees of freedom (d.f.) = 99; $P = 0.53$) and gender ($\chi^2 = 0.38$; d.f. = 1; $P = 0.54$), nor did they differ in age ($t = -0.70$; d.f. = 69; $P = 0.49$) and gender ($\chi^2 = 0.09$; d.f. = 1; $P = 0.77$) of the ill child and type of diagnosis of the ill child ($\chi^2 = 1.7$; d.f. = 3; $P = 0.63$).

The ill child had died in four families at the time of the second measurement (M2). Of the remaining 52 families, three families refused further participation. This resulted in a study group of 49 families (88%) at M2. This group consisted of 66 siblings, 26 boys (39%) and 40 girls (61%), aged 7–18 years (Table 1). There were no significant differences between the M2 non-participant group and the M2 participant group regarding sibling age ($t = 0.04$; d.f. = 81; $P = 0.97$). Although more girls than boys participated in M2 than in M1, this difference failed to reach significance ($\chi^2 = 3.5$; d.f. = 1; $P = 0.06$). Regarding the ill child, no differences were found in age ($t = -0.63$; d.f. = 69; $P = 0.53$) and gender ($\chi^2 = 1.84$; d.f. = 1; $P = 0.18$) between the measurement groups. In the present study, the diagnosis of the ill child is dichotomised into either solid or brain tumours or leukaemia or lymphoma. A significant difference occurs in diagnosis between the M1 and M2 study group ($\chi^2 = 8.5$; d.f. = 3; $P = 0.04$). Of the families who refused to participate at M2, or whose child had died, the ill children were all diagnosed with either a solid tumour ($n = 5$) or a brain tumour ($n = 2$). There were no children with leukaemia or lymphoma in this group. The aforementioned differences according to diagnosis and sibling gender between M1 and M2 resulted in a relative overrepresentation of female siblings with a brother or sister with leukaemia or lymphoma participating at M2 compared with M1.

Table 1a
Participants

Total *N*	Group	Measurement 1	Measurement 2
		n (%)	*n* (%)
Families			
Participants		56 (79)	49 (88)
Excluded	Deceased	–	4 (7)
	Refused	15 (21)	3 (5)
	Total	71	56
Siblings			
Gender	Boys	37 (45)	26 (39)
	Girls	46 (55)	40 (61)
Age (years)	7–12	59 (71)	42 (64)
	13–18	24 (29)	24 (36)
	Total	83	66
Ill child			
Diagnosis	Leukaemia	12 (21)	12 (24)
	Lymphoma	13 (23)	13 (27)
	Solid tumour	26 (46)	21 (43)
	Brain tumour	5 (9)	3 (6)
	Total	56	49

Table 1b
Ill child's mean number of hospital visits according to diagnosis

Period:	Diagnosis–Measurement 1	Measurement 1–Measurement 2
	Mean (S.D.)	Mean (S.D.)
Leukaemia	17 (7)	20 (25)
Lymphoma	28 (9)	33 (13)
Solid tumour	21 (10)	27 (20)
Brain tumour	12 (12)	9 (11)

S.D., standard deviation.

2.2. Procedure

Parents and siblings were approached by letter 4–8 weeks after the diagnosis of cancer in the ill child. After informed consent was obtained, the parents were telephoned and an appointment was made for an interview at their home. The questionnaires were sent in advance by mail, with the instruction to complete the questionnaires alone and independently from other family members. The family was visited for an interview with the parents and the sibling(s) separately, and the completed questionnaires were collected.

The second measurement took place 6 months after the first diagnosis in the ill child. At 6 months, the families were approached by telephone, and were asked whether they wanted to continue participation. When informed consent was prolonged, questionnaires were sent to the siblings and the parents by mail, and an appointment was made for an interview by telephone with one of the parents. Parents and siblings were asked to return the completed questionnaires by mail in a prepaid envelope.

2.3. Measurements

2.3.1. Dependent variables

The *Youth Self Report* (YSR) [32] was used to assess general emotional and behavioural functioning in children aged 11–18 years and is based on the Child Behavior Check List/4-18 (CBCL) (32). The YSR was used in a Dutch version [33]. The YSR yields scores for total behaviour problems, internalising and externalising problems. Scores can be compared with those of a healthy Dutch population of 1016 children aged 11–18 years (495 boys and 521 girls).

Additionally, the *Dutch Children's AZL/TNO Quality of Life Questionnaire* (DucatQoL) (Dr. H. M. Koopman, Leiden University Medical Center, The Netherlands) was used to measure the perception of children aged 7–15 years on daily functioning. The DucatQoL consists of 25 items, scored on a five-point scale. The questionnaire assesses four domains: home, physical, emotional and social functioning. A total quality of life (QoL) score can be obtained. All scores on the subscales are transformed into a scale of 0–100, with higher scores representing a better QoL. Results can be compared with a healthy Dutch norm group stratified by age.

The *State-Trait Anxiety Inventory for Children* (STAI-C) [34] was used in the Dutch translated version for children aged 8–15 years (ZBV-K) [35]. The 'trait' version was used only, assessing the tendency to respond with anxiety to a threatening situation. Dutch norms are available for four separate groups: boys and girls in elementary school ($N = 320$ and $N = 323$, respectively) and in high school ($N = 276$ and $N = 310$, respectively) [34].

2.3.2. Independent variables

Demographic characteristics of the siblings (age and gender), the family (socio-economic status) and the ill children (age and gender) were assessed during the interview with the parents. Information on the ill child's cancer diagnosis was obtained from the medical record of the ill child. The *diagnosis* was dichotomised into either a solid or brain tumour, or leukaemia or lymphoma.

Sibling functioning before diagnosis was reported by the parents retrospectively during the interview at the first measurement. Firstly, the parents were asked whether any physical or psychosocial problems had occurred in the sibling for which a healthcare professional had been consulted (medical or psychosocial) in the year previous to the diagnosis of the ill child. A dichotomous score for the presence or absence of *healthcare use* before diagnosis was computed accordingly. Secondly, the sibling's *physical functioning* prior to the diagnosis of the ill child was assessed during the interview. Parents were asked for any physical, eating or sleeping problems that had occurred during the 2 months preceding the diagnosis in the ill child. Examples of physical problems are: headaches, stomach aches, nausea, vomiting or other undefined problems. Examples of sleeping problems were: trouble falling asleep, problems staying asleep, nightmares, bedwetting or other sleeping problems. Examples of eating problems are: eating too little, or too much, being fussy about food or other eating problems. Scores of physical functioning were obtained on a four-point scale for each complaint from never to monthly, weekly or daily. Subsequently, these three types of physical complaints were summarised in one single dichotomous score for the presence or absence of functional physical complaints before the diagnosis of the ill child.

Parents also scored the major *family life events* that had occurred during the year before the diagnosis of the ill child, from a list of 18 life events. Examples of life events are: the birth of a child; parental divorce; moving; death of a family member, grandparent, other relative or friend; change of school; the termination of a friendship; physical handicap in another child or in a parent; decline in financial means. The presence or absence of at least two major life events prior to the diagnosis of cancer was scored on a dichotomous scale.

2.4. Statistics

Mean scores on social–emotional functioning at M1 and M2 were compared with the norms of each questionnaire, using student's *t*-tests, in order to describe the siblings' functioning at 1 month and 6 months after the diagnosis of cancer in the ill child.

Verhulst and colleagues [33] introduced clinical and borderline cut-off scores for the YSR that corresponded with impaired functioning based on a clinical population. These cut-off scores are used in the present study

to distinguish between siblings who would need professional psychosocial care and those who would not. YSR impaired functioning scores correspond with the 16–17% highest scores in the healthy norm group aged 11–18 [33].

In order to examine whether significant changes occurred in social-emotional functioning over time, paired samples *t*-tests were performed with mean values of behavioural and emotional problems, anxiety and QoL domains of M1 and M2.

To predict functioning 1 and 6 months after diagnosis with previous functioning, and with the ill child's diagnosis, simultaneous regression analyses were performed with M1, M2, and with the difference between these measurements, for boys and girls separately, for all measures of psychosocial functioning. The number of predictor variables was limited due to the relatively small sample of siblings. Therefore, a maximum of one independent variable for each 10 respondents was used as a 'rule of thumb', to ensure sufficient statistical power. The regression analyses were thus conducted with the predictor variables age, previous functioning (physical complaints, healthcare use and life events) and the ill child's diagnosis.

Considering the small number of patients included in the present study and its explorative nature, a significance level of 5% was applied in all analyses.

3. Results

3.1. Prediagnosis functioning

Almost half of the siblings (48%) experienced two or more life events in the year before the illness, as reported by their parents (Table 2). Life events that were mentioned most were: a parent started to work more ($n=16$); a grandparent died ($n=16$); another family member died ($n=13$); a friend of the family died ($n=13$); a parent changed jobs ($n=11$); a parent was diagnosed with a physical handicap ($n=8$); a parent was fired ($n=6$); birth of a child ($n=6$); moving ($n=6$); another child was diagnosed with a physical illness ($n=3$). Furthermore, almost half of the siblings (48%) had used a form of physical or mental healthcare during the year before the diagnosis of cancer in a brother or sister. As to problems in overall daily functioning, parents reported that one or more physical, sleeping or eating problems had occurred in 52% of the siblings. These problems were either physical (35%), sleeping problems (27%) or eating problems (21%) or a combination of these three physical domains.

Table 2
Frequencies of problems before diagnosis

	Life events (⩾2)	Healthcare use (⩾1 month)	Health problems (⩾1)			
			Total	Physical	Sleeping	Eating
%	47	47	52	35	27	21
N	39	39	43	29	22	17
Mean (S.D.)	1.66 (1.38)	4.15 (6.83)	2.16 (2.92)	0.82 (1.42)	0.83 (1.61)	0.51 (1.06)

3.2. Social-emotional functioning 1 month after diagnosis

Adolescent girls reported significantly more internalising problems (YSR) and anxiety (STAI-C) than their peers at M1 (Table 3). Both children (7–11 years) and adolescents (12–18 years) reported significantly impaired emotional, social and overall QoL, compared with the normal population. In addition, 7–11-year-old siblings reported impaired physical QoL compared with the normal population.

Furthermore, the percentage of siblings having scores in the borderline or clinical range was considered for the YSR. A significant number of female adolescent siblings reported internalising (48%; $\chi^2=14.32$; $P<0.0001$), externalising (33%; $\chi^2=4.35$; $P<0.05$) and overall problems (43%; $\chi^2=10.32$; $P<0.01$) in the borderline or clinical range of the YSR, compared with the percentages in the normal population (16, 16 and 17%, respectively). The number of adolescent boys who reported problems in the clinical range was less substantial (32, 11 and 21%, respectively) and not significantly different than that in the normal population (17% for all three scales).

3.3. Sibling social-emotional functioning 6 months after diagnosis

At M2, the QoL of the *children* was still somewhat, but not significantly, lower than that of the normal population of children. The emotional, social and total QoL of the *adolescents* was still significantly lower compared with the normal population of adolescents. Adolescent girls still reported high levels of internalising problems (YSR) and anxiety (STAI-C), but not significantly more than the normal population. Approximately 35% of the girls still reported internalising and externalising problems in the clinical borderline range of the YSR, which is significantly more than in the normative group for internalising ($\chi^2=4.41$; $P<0.05$) and externalising problems ($\chi^2=4.41$; $P<0.05$).

Boys in elementary school (STAI-C) reported lower anxiety scores than their peers at M2. Adolescent boys reported lower externalising and total behaviour problems (YSR) than their peers at M2 (Table 3).

3.4. Changes in functioning 1–6 months after diagnosis

Paired *t*-tests showed that adolescent boys reported significantly fewer externalising behaviour and total problems (YSR) on M2 than on M1 (Table 3).

Table 3
Means of social-emotional functioning one (M1) and six (M2) months after diagnosis and norms

	NORM			M1			M2			M1−M2	
	N	M	S.D.	N	M	S.D.	N	M	S.D.	Mean difference	95% Confidence interval of the difference
YSR[a]											
Internalising											
Boys	495	8.3	5.7	19	9.3	7.8	15	6.5	6.4	2.23	−1.13 to 5.58
Girls	521	10.6	6.9	21	16.8*	10.0	17	15.1	12.3	0.63	−5.14 to 6.40
Externalising											
Boys	495	11.2	6.4	19	10.5	9.6	15	7.2*	9.0	2.77**	1.33 to 4.21
Girls	521	9.8	5.9	21	11.3	7.8	17	10.5	7.3	0.69	−2.42 to 3.80
Total											
Boys	495	32.8	16.3	19	33.5	22.9	15	23.4*	21.5	7.10**	3.65 to 10.56
Girls	521	33.9	17.3	21	46.9	26.1	17	40.3	26.5	4.47	−7.60 to 16.54
STAI-C[b]											
Child											
Boys	320	31.2	5.5	24	30.8	7.9	11	24.0**	3.3	4.25	−1.75 to 10.25
Girls	323	33.4	6.5	31	33.3	7.6	24	30.1	7.0	2.69**	1.17 to 4.22
Adolescent											
Boys	276	28.7	6.4	11	31.9	7.7	9	30.3	9.6	2.25	−5.09 to 9.59
Girls	310	32.5	6.7	12	36.5*	9.7	12	33.5	9.0	1.63	−4.87 to 8.12
DucatQoL[c]											
Child (elementary school)											
Home	880	86.5	13.9	45	83.4	13.9	30	87.1	15.2	−1.89	−7.68 to 3.91
Physical	873	79.4	15.6	45	72.6*	21.4	29	79.0	18.1	−3.20	−10.37 to 3.97
Emotional	877	74.4	15.5	45	61.9**	18.5	30	69.9	19.6	−4.96	−12.47 to 2.54
Social	871	79.1	13.4	44	73.2**	15.7	28	74.5	15.1	−1.16	−8.41 to 6.08
Total	838	79.6	12.2	44	71.8**	13.7	27	76.4	14.4	−4.10	−10.62 to 2.42
Adolescent (secondary school)											
Home	262	77.4	17.9	35	75.8	17.2	23	72.0	19.3	1.18	−4.94 to 7.29
Physical	266	64.9	21.1	35	58.9	22.5	24	59.3	18.3	−0.61	−6.59 to 5.37
Emotional	265	67.7	16.7	34	59.7*	18.9	24	59.6*	16.0	−1.16	−7.42 to 5.10
Social	254	73.6	12.6	34	64.9**	13.4	24	65.4**	15.0	−2.57	−8.14 to 3.00
Total	251	70.8	13.1	34	64.3**	15.3	23	63.0**	14.9	−0.54	−5.30 to 4.22

M1, measurement 1; M2, measurement 2; YSR, Youth Self Report; STAI-C, State-Trait Anxiety Inventory for Children; DucatQol, Dutch Children's AZL/TNO Quality of Life Questionnaire.

*$P<0.05$; **$P<0.01$.

[a] YSR: high scores represent more social–emotional problems.

[b] STAI-C: high scores represent more anxiety.

Elementary school-aged girls reported a significant decrease in anxiety at M2 compared with M1 (Table 3). QoL of the siblings (both children and adolescents) increased, but not significantly.

3.5. Predictors of functioning

Regression analyses with the predictor variables were conducted for M1 and M2 and for the difference between measurements, for boys and girls separately. No significant effects were found for any of the predictors of the mean difference scores between M1 and M2. Therefore, only the results for M1 and M2 are presented (Table 4a and b).

At the *first measurement*, age was a significant predictor for QoL in all domains except for emotional QoL in the girls. The older, the more impaired the sisters' QoL. Life events were also a predictive factor for girls. Sisters that had experienced several major life events before their brother or sister became ill seemed at risk for impaired functioning in the family, in their social relationships and in their overall functioning. In addition, girls whose brother or sister was diagnosed with leukaemia or lymphoma reported more impaired physical, social and total QoL than girls whose brother or sister was diagnosed with a solid or brain tumour. In the boys, none of the variables appeared to predict their social–emotional functioning at 1 month after diagnosis.

At the *second measurement*, age was a significant predictive factor of sibling functioning on several domains:

Table 4
(a) Simultaneous regression analyses at M1 with predictor variables; (b) Simultaneous regression analyses at M2 with predictor variables (standardised βs)

	YSR			STAI-C	QoL				
	Int	Ext	Tot	Anxiety	Home	Phys	Emo	Soc	Tot
(a)									
Boys									
Regression model									
R^2	0.21	0.26	0.20	0.09	0.05	0.18	0.18	0.28	0.21
F	0.68	0.93	0.64	0.56	0.29	10.24	10.22	20.12	10.40
(β)									
Age	0.20	0.29	0.31	0.07	−0.22	−0.24	−0.14	−0.27	−0.27
Life events	0.39	0.20	0.33	0.13	0.00	−0.23	−0.23	−0.04	−0.18
Healthcare use	0.14	−0.20	−0.07	0.10	−0.02	−0.12	−0.10	0.06	−0.07
Physical functioning	0.08	−0.29	−0.18	0.27	0.04	0.06	−0.03	0.30	0.12
Leukaemia/lymphoma	0.17	−0.32	−0.11	−0.03	−0.01	0.22	0.33	0.20	0.26
Girls									
Regression model									
R^2	0.33	0.30	0.30	0.11	0.31	0.47	0.17	0.33	0.40
F	10.45	10.28	10.26	0.92	30.39*	60.76**	10.55	30.59*	40.93**
(β)									
Age	0.32	0.26	0.28	0.19	−0.41*	−0.71**	−0.25	−0.31*	−0.52**
Life events	−0.04	0.13	0.04	0.15	−0.41*	−0.22	−0.28	−0.34*	−0.37**
Healthcare use	0.18	0.40	0.30	0.21	0.13	0.09	0.08	0.00	0.09
Physical functioning	−0.04	0.03	−0.01	−0.09	0.04	0.09	0.23	−0.10	0.09
Leukaemia/lymphoma	0.59*	0.37	0.51	0.07	−0.23	−0.36*	−0.16	−0.45**	−0.37*
(b)									
Boys									
Regression model									
R^2	0.46	0.23	0.37	0.32	0.34	0.47	0.50	0.50	0.57
F	10.52	0.55	10.07	10.29	10.43	20.53	20.97*	30.04*	30.46*
(β)									
Age	0.63	0.36	0.60	0.52	−0.60*	−0.60*	−0.63*	−0.52*	−0.65**
Life events	−0.33	−0.21	−0.27	−0.28	0.19	−0.16	−0.03	0.02	−0.02
Healthcare use	0.20	−0.23	−0.06	0.09	−0.08	0.02	0.10	0.20	0.13
Physical functioning	0.51	−0.08	0.16	0.07	−0.28	−0.21	−0.20	−0.32	−0.18
Leukaemia/lymphoma	−0.04	−0.02	−0.08	−0.13	0.14	0.35	0.23	0.32	0.23
Girls									
Regression model									
R^2	0.19	0.12	0.20	0.11	0.32	0.47	0.18	0.07	0.31
F	0.53	0.30	0.54	0.72	20.56	40.77**	10.15	0.39	20.23
(β)									
Age	0.29	0.16	0.19	0.17	−0.41*	−0.61**	−0.32	−0.21	−0.43*
Life events	0.05	0.16	0.03	0.25	−0.30	−0.06	0.13	−0.04	−0.15
Healthcare use	−0.02	0.10	0.14	0.08	0.13	0.16	0.03	0.07	0.22
Physical functioning	−0.20	−0.01	−0.09	0.02	−0.17	−0.17	−0.02	−0.12	−0.22
Leukaemia/lymphoma	0.36	0.19	0.39	0.09	−0.10	−0.35*	−0.32	−0.14	−0.20

Values are standardised regression coefficients (β) with significance of t, except for the Regression Model where rows represent R^2 and F values and significance of F is displayed. *$P<0.05$ **$P<0.01$.

M1, measurement 1, 1 month after diagnosis; YSR int, Youth Self Report, Internalising problems; YSR ext, Youth Self Report, Externalising problems; YSR tot, Youth Self Report, Total problems; QoL, Quality of Life DucatQoL; home, phys(ical), emo(tional), soc(ial), tot(al); YSR, Youth Self Report; STAI-C, State-Trust Anxiety Inventory for Children; DucatQol, Dutch Children's AZL/TNO Quality of Life Questionnaire.

the older the boys, the more impaired their emotional, social and total QoL, the older the girls, the more impaired their physical QoL. Girls whose sibling was diagnosed with either leukaemia or lymphoma still reported relatively more impaired physical functioning at M2 compared with girls whose brother or sister was diagnosed with a solid or brain tumour.

4. Discussion

The object of this study was to examine the psychosocial functioning of siblings of paediatric cancer patients at 1 and 6 months after diagnosis and to investigate the risk factors that explain why some siblings are more at risk for these problems than others.

In earlier studies on sibling psychosocial functioning, the factor time was rarely included, and considerable variation in study designs resulted in contradictory findings.

From the present study results, it appeared that siblings were at risk for impaired psychosocial functioning and impaired QoL, shortly after diagnosis. The severity of psychosocial distress in the siblings seemed to diminish over time, but impaired QoL remained present in subgroups of siblings in the first 6 months after diagnosis. Several factors seemed to play a role in the level of psychosocial distress experienced during this stressful period.

First, differences between children and adolescents became clear when the sibling's QoL is compared with normative data. In the children, the first distress seemed to translate itself into negative evaluations for emotional, physical and social QoL. In adolescent siblings, problems manifested in impaired emotional QoL and in social and overall QoL, but not in impaired physical functioning. Considering their cognitive developmental level, children are less capable to understand why and how their sibling got ill. They may be relatively frightened of getting ill themselves, which may be expressed in physical complaints.

After six months, the children's QoL seemed to have normalised when compared with a normative peer group. However, problems in social, emotional and overall QoL seemed to persist in adolescent siblings. Adolescents still experienced problems in daily functioning at 6 months after diagnosis.

When the emotional and behavioural reactions of siblings were compared with normative data, differences between boys and girls became apparent. Shortly after diagnosis, adolescent sisters reported more internalising behaviour such as anxious–depressive symptoms, emotional withdrawal and physical complaints than their peers. These internalising problems seemed to normalise over time. Male siblings did not report more internalising problems than peers and reported even fewer externalising, behavioural and emotional problems than their peers at 6 months after diagnosis. Although this trend extended over different questionnaires and age groups, these results must be interpreted cautiously because they are based on very small subgroups of boys.

When the predictive factors for sibling psychosocial functioning were considered, again adolescent siblings appeared to be a risk group in the first 6 months after diagnosis. On several domains, depending on their gender, they reported more impaired QoL. Girls reported more impaired QoL shortly after diagnosis with increasing age. The older they were, the more impairment they experienced in their relationships with others and with family members and in their physical and overall functioning. Six months later, both girls and boys seemed to report more impaired functioning with increasing age, but on different domains of functioning. With increasing age, sisters seemed most vulnerable for physical problems, whereas brothers suffered most from emotional and social problems and also showed more impaired overall functioning with increasing age. These social and emotional problems and problems in overall functioning in the brothers corresponded with the domains where the adolescent siblings experienced impaired QoL when compared with peers.

In conclusion, adolescent siblings seemed to be especially vulnerable for increased psychosocial problems. They were affected longer and more seriously by the illness than the children in this study and seemed to experience problems in specific domains, depending on their gender. An explanation from a developmental psychological perspective seems most appropriate. From a cognitive viewpoint, adolescents may develop the ability to hypothesise about the course of events. They have the capability to estimate probabilities and limitations in a realistic way. In addition, adolescents develop an irreversible consciousness about their own mortality, which, especially in siblings, can lead to existential fears. Consequently, adolescent siblings are fully capable of estimating and understanding the present risks and future consequences of their sibling's illness. In the same developmental phase where existential fears appear, this may result in an increased emotional vulnerability, resulting in psychosocial adjustment problems. Adolescents are already more vulnerable for internalising problems, such as sadness, depression, anxiety and emotional withdrawal [36]. The present results corresponded with the hypothesis that the intense concern and responsibility for the patient and the parents may interfere with their developmental task of separation and individuation [2]. The occurrence of a life-threatening illness in a brother or sister may thus be more intrusive for adolescent siblings who understand more, are relatively emotionally vulnerable and are curbed in their social and emotional development by the illness experience.

Besides age, two other factors predicted the sisters' QoL in particular: life-events and the ill child's diagnosis. When several life-events have already burdened the family before the illness, female siblings were especially vulnerable for distress experienced in relationships with family members and other social relationships in the first month after diagnosis. This effect did not persist over time. For the girls, more problems were reported by those who had a brother or sister with leukaemia of lymphoma, compared with solid or brain tumours. Lower satisfaction with their own physical functioning was the most persistent problem in these girls over time. This effect may be explained by the longer and more intensive treatment for leukaemia or lymphoma. The more intensive treatment for leukaemia or lymphoma may have led to an increased separation from the parent and to more responsibilities. This greater burden may be expressed in the relatively higher physical complaints in this subgroup. That this effect was only present in the

girls may be due to an over-representation of children with leukaemia or lymphoma in the subgroup of girls. Siblings who needed healthcare use or who already experienced physical problems before diagnosis did not appear to be more vulnerable for social–emotional problems of any kind over time.

Several comments should be made on the aforementioned results. Firstly, an involuntary selection of families according to the diagnosis of the ill child occurred after the first measurement. This resulted in a non-representative group with a slight over-representation of ill children diagnosed with leukaemia or lymphoma, with better prognoses in the second measurement group. Secondly, the numbers of siblings in the subgroups that were stratified by age and gender were relatively small, especially in the subgroups of boys. Therefore, effects of predictor variables do not reach significance easily, and effects may have differed by chance between boys and girls. Thirdly, the amount of variance explained by age, previous life events, previous health care use, previous functioning and diagnosis of the ill child was low. Besides, the high prevalence of previous problems in the siblings reduced the predictive power of this variable.

None of the selected variables explained the changes in functioning over time. This indicates that other factors, such as coping, family functioning and communication about the illness, might play a more prominent role and need to be investigated in the future. In spite of these limitations, the persistence of problems in subgroups of siblings underlines the importance of longitudinal follow-up. Adolescent siblings seem to need extra attention and social support. Positive effects have been obtained with supportive groups for siblings in different phases of the illness [37–39]. Sibling's QoL may be protected or even ameliorated in the first six months when they are offered extra support in a peer group of other siblings who have had similar experiences. Internalised feelings of anxiety, anger, jealousy and loneliness can be ventilated and worries can be shared with their peers. This approach may prevent later problems.

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